Perineal endometriosis in an episiotomy scar: case report and review of literature

Teresa Tam, Stella Huang
Department of Obstetrics & Gynecology, Resurrection Health Care/Saint Joseph Hospital, Chicago, Illinois - USA

ABSTRACT
Purpose: Perineal endometriosis is a rare disease characterized by the presence of ectopic endometrial stroma and glands in the perineum. Most commonly observed in the episiotomy scar after normal vaginal deliveries, perineal endometriosis is not often considered in the differential diagnosis of perineal masses. This could lead to unnecessary investigations and inadequate patient treatments.
Case Report: A 41-year-old woman complains of a tender perineal mass, which becomes hard and more painful during menses. Her symptoms started five years prior in the same area where a midline episiotomy was performed during a normal vaginal delivery. Complete surgical excision of the perineal mass was performed and the pathology report confirmed perineal endometriosis. No recurrence was noted on the postoperative course.
Conclusion: A comprehensive history and meticulous pelvic examination are essential in diagnosing perineal endometriosis. Surgical intervention is the best approach for treatment and permanent cure is usually achieved after complete excision of the perineal endometriosis.

KEY WORDS: Endometriosis, Episiotomy, Perineal endometriosis, Perineal mass
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INTRODUCTION

Endometriosis is a benign disease defined by the presence of endometrial glands and stroma in anatomic locations other than the uterine lining. It is most commonly observed in the dependent portions of the pelvis, and most frequently on the peritoneal surfaces of the ovaries, uterosacral ligaments, and other pelvic organs. Endometriosis can also be found anywhere in the body such as the thoracic cavity, urinary and intestinal tracts, inguinal canal, umbilicus, surgical scars, and the perineum.

The incidence of perineal endometriosis is rare, possibly because of underreporting. Its significance in clinical practice is arguable. In a review of the literature, only case reports and case series have been reported. Between 1983 and 2007, the incidence of perineal endometriosis was 0.31% among women with endometriosis treated surgically at Peking Union Medical College (1). The rarity of cases may be because of the fact that perineal endometriosis may not have been included in the differential diagnosis of perineal masses, which may have led to underreporting and inadequate patient treatment.

Case Report
A 41-year-old Asian woman (G2P2002), presented with complaints of perineal discomfort and a fluctuant perineal mass, which became hard and more painful during menses. Her symptoms commenced five years prior, in the same area where a midline episiotomy was performed during a normal vaginal delivery. She had refrained from sexual intercourse since her last delivery because of se-
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A 0.5 cm, bluish, circumscribed, indurated, and tender mass, visible upon inspection of the perineum. This mass was located midline, halfway between the introitus and the anus, in the area of the perineal episiotomy scar. A photograph was not obtained so a graphic illustration is shown in Figure 1. Pelvic and rectal examinations were normal.

With clinical history and physical findings alone, perineal endometriosis was strongly suspected. Because of the small size of the lesion and no suspicion of anal sphincter involvement, radiologic imaging or laboratory testing were not obtained. The patient was informed that surgical excision was the best choice of treatment. After extensive discussion of the procedure, the patient was scheduled for total excision of the perineal mass.

Under general anesthesia, the mass was completely excised together with deep surrounding healthy tissue. This ensured that no residual endometriosis was left behind. A chocolate cyst with multiple loculations, measuring 0.75 cm in diameter, was sent to pathology. Perineorrhaphy repair with absorbable sutures was performed. Histologic examination of the surgical specimens revealed perineal tissue with endometriosis consisting of both endometrial glands and stroma (Figs. 2 and 3).

In the post-operative period, the perineal incision site healed and approximated well. The patient reported no perineal discomfort during or after menses. No recurrence was noted during the post-operative course. The patient remained asymptomatic one year later and has resumed normal sexual activity.

Fig. 1 - Location of perineal endometriosis (A) in episiotomy scar.

Fig. 2 - Microscopic appearance of perineal endometriosis in episiotomy scar, showing endometrial glands and stroma. (Hematoxylin & Eosin stain, 10x).

Fig. 3 - Higher magnification of perineal tissue with endometriosis. (Hematoxylin and Eosin stain, 40x).
DISCUSSION

The incidence of endometriosis is 7% to 10% (2). Although endometriosis is a common disease in women, there are only sporadic cases of perineal endometriosis reported in the gynecology literature. Reported cases arise mostly at the site of previous episiotomy scars, perineal trauma or after pelvic radiotherapy (3). Few cases occur without prior perineal trauma (4). Pollack et al (5) were among the first to report this in 1990. In 2008, a case of perineal endometriosis in pregnancy was reported in a previously diagnosed case, which had poor follow-up (6). The endometriotic lesion subsequently grew in size to 5 cm, causing severe pain during the third trimester, which led to cesarean delivery. Extensive perineal endometriosis can extend to the anal sphincter, with six reported cases in 1991 (7), requiring surgical excision plus partial sphincteroplasty. Because of rare occurrences of perineal endometriosis and their potential for extensive involvement, reporting all cases is important.

The diagnosis of perineal endometriosis is based mainly upon the specific clinical manifestations and characteristic local findings in physical examination. A detailed history and thorough perineal and pelvic examination ensure correct diagnosis, which leads to early treatment and quick relief of symptoms. A tender nodule or perineal mass, accompanied with progressive and cyclic perineal pain in a patient with a history of an episiotomy, is highly diagnostic (8). Physical examination usually reveals a tender bluish perineal mass. Previously reported cases consistently describe these physical exam findings. Symptoms usually appear shortly after ectopic endometrial cell implantation, with some cases having a prolonged latent period of up to 14 years after implantation (9).

This direct implantation theory could explain the occurrence of perineal endometriosis after gynecologic procedures. Uterine curettage, manual uterine exploration or uterine inversion may increase the incidence of perineal endometriosis (10,11). Some studies have shown that lochia from curettage contained a higher number of decidual cells, endometrial cells and glands, and scattered syncytial trophoblastic cells, compared to lochia obtained after delivery (12). Cells from the zona basalis likely attach to the fresh wound of an episiotomy or perineal laceration and become trapped during healing. The mass subsequently continues to grow to a size that causes discomfort and pain, especially during menses.

Paull and Tedeschi, in exploring this theory, found a higher incidence of perineal endometriosis in women having curettage after an episiotomy, in comparison to women who only had an episiotomy (12). In the few cases of perineal endometriosis without perineal trauma (4,5), endometrial cells may have been lymphatically disseminated through the uterus, cervix, vagina, and perineum. This is the theory behind extraperitoneal endometrial foci including the lungs, pleura, skin, skeletal muscle, and central nervous system (13). In rare cases, benign endometriosis can undergo malignant transformation after several years (14). Malignant degeneration of extragonadal endometriosis occurs in 21.3%, with 0.3% to 1% occurring in surgical scars (15).

Serum CA-125 glycoprotein antigen can also be used to diagnose endometriosis but only has sensitivity ranging between 13% to 36% (16). Women with endometriosis often have greater than 35 IU/ml serum CA-125 concentrations. It is not, however, a sensitive indicator of endometriosis. The best correlation is seen in stage III or IV disease (17). Concentrations of CA-125 can also be elevated in epithelial ovarian carcinoma, endometrial cancer, pelvic inflammatory disease, and adenomyosis, among other conditions. In one study, serum CA-125 levels were elevated (>35 IU/mL) in two out of 31 cases (6.5%) that had perineal endometriosis with anal sphincter involvement (18). These cases also had ovarian endometriomas. Thus, its utility in diagnosing perineal endometriosis may only be useful in more severe cases with extensive involvement that may suggest pelvic endometriosis.

Perineal endometriosis can be diagnosed by means of symptoms and radiologic imaging. Ultrasound can characterize masses, revealing endometriotic implants’ internal characteristics, depth of invasion, size, and their relationship to surrounding structures. These masses typically show an echo-complex consistency with irregular outline (19) and heterogeneous hyperechoic images, possibly distinguishing them from a malignancy.

Radiology literature endorses the role of endorectal sonography in assessing sphincter involvement prior to surgical treatment of perineal endometriosis so that complete resection may be achieved. Pelvic MRI shows greater sensitivity (90% to 92%) and specificity (91% to 98%) for diagnosing endometriomas compared to CT and ultrasound (9).

The treatment of perineal endometriosis includes surgical intervention and medical manipulation by means of hormonal suppression. The first choice is complete excision of the perineal endometrial tissue because it offers both diagnostic and therapeutic opportunities. It is the most effective procedure for permanent cure in young
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and healthy patients and is mandatory, especially if malignancy is suspected. If residual lesions are noted post-operatively, cauterization and hormonal therapy are indicated. Incomplete removal predisposes the patient to recurrence of endometriosis, which often occurs within one year after treatment (20).

Hormonal suppression with medroxyprogesterone, testosterone or danazol attempts to interrupt the cycles of stimulation and bleeding of endometriotic tissue by interrupting cyclic ovarian hormone production (21-23). Some clinicians with varying degrees of success investigated this. The major disadvantage of medical treatment is that perineal endometriosis often persists. Although symptomatic relief could be achieved with hormonal intervention, complete surgical excision still remains the best treatment for perineal endometriosis and often leads to permanent cure.

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Address for correspondence:
Maria Teresa Tam
Department of Obstetrics & Gynecology
Resurrection Healthcare/ Saint Joseph Hospital
2900 North Lake Shore Drive
Chicago, Illinois 60657
USA
maria.tam@reshealthcare.org

REFERENCES